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Crohn's disease of the vulva

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Keywords: Crohn's disease; vulva; ulcerative colitis

Cutaneous manifestation of Crohn's disease may appear sometime after gastrointestinal presentation. To our knowledge this is the only case reported where the vulval lesion did not appear until 20 years after the initial diagnosis had been made. The patient had suffered no cutaneous involvement in the intervening years.

Case report

A 59-year-old married woman was referred to the gynaecological outpatient department with a one year history of left sided vulval swelling and tenderness.

She had a past history of ulcerative colitis which had been diagnosed on rectal biopsy in 1967. This was initially controlled by salazopyrin therapy but a series of severe exacerbations of the disease resulted in her having a total colectomy and ileostomy in 1973. Following this, the disease remained quiescent and she was discharged from the surgical clinic on no treatment in 1981. She had been well since.

On examination she appeared pale. Abdominal examination revealed a functioning ileostomy. There were no palpable masses. There was swelling and oedema of the left labium majus which was twice the size of the right (Figure 1). She had a grossly hypertrophied clitoral hood with several superficial ulcers on the skin around the clitoris, perianal region and lateral margins of the labium majus, some of which were infected. The entire skin surface was reddened and inflamed. The vaginal epithelium was atrophic and there were thin filmy adhesions in the vaginal vault. The uterus and adnexae were normal.

Two biopsies were taken. Histology revealed hyperplasia of pseudocarcinomatous appearance. The specimen contained profuse numbers of chronic inflammatory cells, predominantly lymphocytes and plasma cells. In addition there were numerous non-caseating granulomata composed of Langhans type giant cells and epithelioid cells. Special stains showed no evidence of acid-fast bacilli. On the result of the histology a diagnosis of Crohn's disease was made. The histopathologist was asked to review the histology of the patient's colectomy specimen, of 14 years previously. The conclusion drawn was that because of the active nature of the colitis it would have been difficult to have made a firm diagnosis at that time. On balance and in the light of subsequent findings it was concluded that a diagnosis of Crohn's disease would have been more likely.

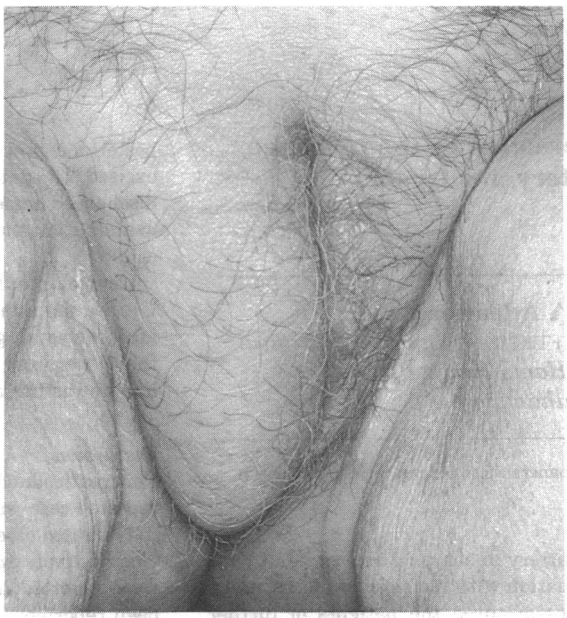

Figure 1. Vulva showing hypertrophied left labium majus

The patient was initially managed with Dermovate NN Cream applied to the vulva thrice daily. When reviewed 2 months later her symptom of pain had improved dramatically. However, on clinical examination the vulva appearance remained the same. She was therefore commenced on metronidazole 400 mg twice daily. After 3 months' therapy there was complete symptomatic and objective healing which has been maintained one year after discontinuation of all treatment.

Discussion

Extensive cutaneous ulcerative Crohn's disease affecting the vulva was first described in 1965[1]. Some authors have now reported incidences of cutaneous manifestations of Crohn's disease to be present in between 22 and 44% of cases[2,3]. Only a few cases of vulval involvement have been reported[4].

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The most common site of cutaneous involvement in Crohn's disease is the perineum[5] with lesions occurring in 43-94% of patients. Internal fistulas such as recto-vaginal[6], ileo-uterine and ileo-tubal[7] have also been reported.

Corticosteroids azathioprine and sulphasalazine have been the mainstay of medical treatment for vulval Crohn's. The results of various local treatments with steroids and/or antibacterial agents have been disappointing[8]. Systemic steroids and cytotoxic agents have also been shown to have no beneficial effect on the vulval lesion. Regular curettage of vulval ulcers with concomitant oral zinc sulphate administration have been used to good effect in some patients[9]. It has also been recommended that removal of the adjacent bowel is of benefit if it is involved in the disease process, as cutaneous ulceration is unlikely to heal when diseased intestine is still present. Surgical intervention is however, followed by unhealed wounds in up to one half of patients[10]. Limited local excision will frequently result in recurrence and failure to control the lesion. Extensive radical excision of all the area involved has been proposed as the best chance of cure[11].

The long-term use of metronidazole for perianal and vulval disease has recently been found to be of use[12]. A standard regimen has been instituted in some units with regard to vulval ulceration. This includes local care in the form of iodine and a dextromononer absorptive dressing. This is combined with prednisolone orally and long term metronidazole therapy[13]. Most patients tolerate metronidazole therapy well without significant side effects. Minor side effects include a metallic taste and darkening of the urine together with gastrointestinal upset. More severe side effects such as peripheral neuropathy may occur in long-term therapy.

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A fatal association between a seizure and drug-induced parkinsonism

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Keywords: seizure; drug-induced parkinsonism; respiratory muscles; dystonic symptoms; epilepsy; fatality

Dystonic and dyskinetic symptoms associated with antipsychotic medication and involving the pharynx and larynx may present a risk for aspiration and/asphyxiation[1,2]. Previously it had been shown that terminal seizures were sometimes associated with sudden death in cases currently receiving antipsychotic medication[3]. We now present a case in which the adverse synergistic effects of a convulsion coupled with a drug-induced parkinsonian syndrome involving the respiratory muscles is highlighted.

Case report

A 19-year-old woman was admitted 7 weeks prior to her death to the general medical ward in a mute catatonic state. She was initially investigated for a possible underlying neurological disorder but this proved negative. She was then given chlorpromazine (100 mg three times a day) plus fluphenazine decanoate intramuscular injections (125 mg/week for 3 weeks following admission). At the end of 3 weeks she was no longer catatonic nor mute but was grossly psychotic and difficult to manage in the general ward. She had also developed severe dystonic side effects, which restricted her breathing when the rigidity was severe. She received biperiden (5 mg intramuscular as required) which controlled her dystonia. Her slow progress led to a cessation of all previous neuroleptic treatment from the 4th week of her stay and clozapine (300 mg nocte) was substituted and the biperiden was continued. Two weeks later (ie 6 weeks from admission) her response remained minimal and flupenthixol intramuscularly (40 mg biweekly) was added to her medication. At the same time biperiden was increased (5 mg intramuscularly daily) because her dystonic reactions continued intermittently. However her general condition was still only ameliorated to a minimal degree. On the day of her death she had a dystonic reaction in the morning which was controlled by a biperiden injection. She recovered fully, ate lunch, but at 1500 h she again became dystonic and suffered a major grand mal convulsion (never having had a fit prior to this) before she could receive further biperiden. Death followed within 15 min, apparently from asphyxia. There was no clinical evidence of aspiration and no post mortem examination was carried out.

Discussion

In this case it was likely that the preceding dystonic reaction was related to the fatal outcome following the fit, in that it could have seriously impaired respiratory function to such an extent that the patient was unable to recover from the fit. It would appear that any epileptic patient on major tranquillizers or any patient suffering with fits while on neuroleptics may well be at greater risk of a fatal outcome from a seizure. It is possible that this could result from the adverse synergism between the drug-induced extrapyramidal symptoms, especially when affecting the respiratory muscles, and a concomitant convulsion. In view of the well known lowering of seizure threshold common to most antipsychotic agents currently in use[4], it might be worthwhile to combine an anti-epileptic agent with such medication on a routine basis particularly if larger than normal doses of neuroleptics are administered. Moreover, in patients who are prone to develop parkinsonian side effects, switching exclusively

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